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Case Report

Dermatology Section

A Case of Facial Discoid Lupus Erythematosus (LE) with Oral Lichen Planus (LP): A Dig into Co-existence and LE-LP Overlap

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ABSTRACT

A distinction between 'co-existence of Lupus Erythematosus (LE) and Lichen Planus (LP)' and 'LE-LP overlap' bears importance as the criterion for true overlap necessitates histological and immunological features of both LE and LP to be found in the same tissue specimen. However, in the present case report the lesions of LE and LP were present in two different sites. A 51-year-old male presented with a large erythematous scaly atrophic plaque on the right cheek with similar lesions on chin and lips for 10 years. He also had violaceous lesions on the bilateral buccal mucosa and a white lesion over the left buccal mucosa for three years. Discoid Lupus Erythematosus (DLE) and Oral Lichen Planus (OLP) were suspected. Dermoscopy of the facial plaque showed features consistent with DLE. Histopathology of the facial plaque confirmed the diagnosis of DLE, whereas the white plaque on the left buccal mucosa showed features of early invasive squamous cell carcinoma. Violaceous lesion over the right buccal mucosa showed features suggesting OLP. A Direct Immunofluorescence (DI) was also performed for the buccal mucosa to rule out the possibility of DLE with oral involvement, which turned out to be negative. Therefore, a diagnosis of synchronous presentation of DLE and OLP along with Squamous Cell Carcinoma (SCC) of the buccal mucosa was made and patient was treated with topical corticosteroids, systemic hydroxychloroquine and surgical intervention for the squamous cell carcinoma. It is also imperative that not only long standing cutaneous lesions, but also oral lesions like LP should be investigated and kept under observation to look for any early malignant changes.

Keywords: Dermoscopy, Direct immunofluorescence, Follicular plugging, Oral squamous cell carcinoma, White rosettes

CASE REPORT

A 51-year-old male patient, painter by occupation presented with a red, scaly and atrophic plaque on the right cheek since past 10 years. Patient had similar lesions on the chin and lips as well. The lesions started as mildly erythematous pruritic patches which gradually evolved over years to reach the current state. There was history of photosensitivity as he complained of burning sensation over the lesion on sun exposure. There was no history of chronic drug intake. There were no lesions elsewhere on the body. There was history of similar lesions in the family.

The patient also complained of multiple bilateral oral lesions over the buccal mucosa for three years. He had pain and irritation while chewing and deglutition. He was a chronic smoker (nine pack-years) and also chewed tobacco for 18 years. He also consumed alcohol (30-60 mL of country liquor twice weekly for past 10 years). There was no history of application of dental amalgam. He did not give any history of systemic co-morbidities or loss of weight or appetite.

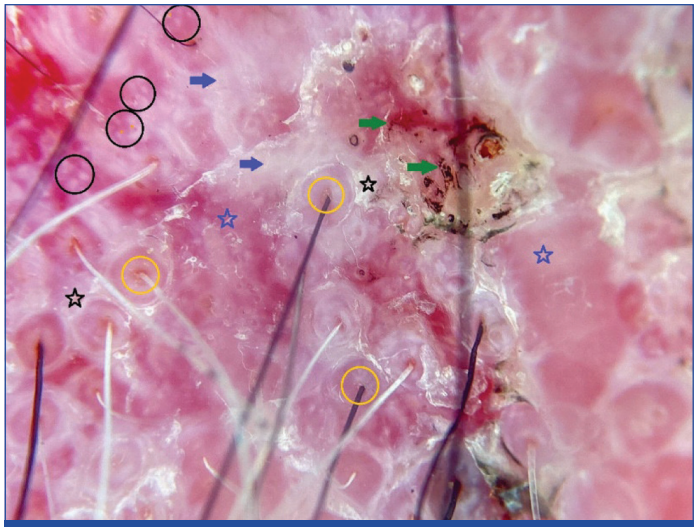
On examination, the plaque over right cheek measured approximately 10×6 cm and extended from below the zygomatic bone upto the chin. It showed prominent erythema, adherent scaling, atrophy and scarring with few areas showing loss of hair. There was central depigmentation and peripheral hyperpigmentation [Table/Fig-1]. The surface was rough and borders were well-defined. No discharge was noted. The lesion did not show warmth or tenderness. Regional lymphnodes were non palpable. There was no growth over the surface. The lesion did not bleed on touch. The lesions on the chin and lips showed similar findings. Based on clinical morphology, a primary diagnosis of DLE was made.

The buccal mucosa showed erythematous and violaceus plaques over bilateral walls. Mild superficial erosions were noted. The tongue, palate and gums were free from involvement. There was poor dental hygiene and tobacco stains on the teeth noted. The plaque on the left buccal mucosa showed development of superficial white adherent patch [Table/Fig-1].

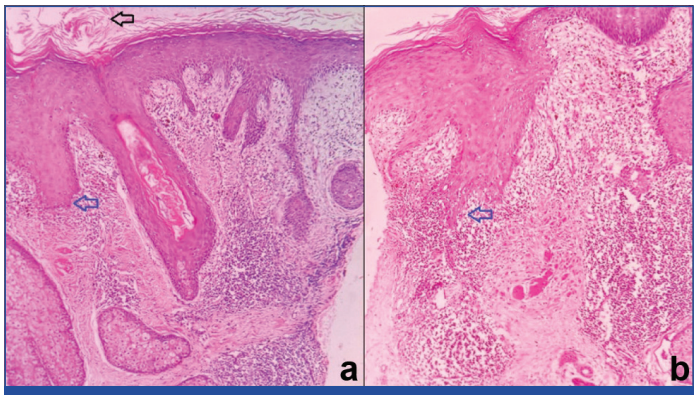
[Table/Fig-1]: a) Clinical image of the facial plaque with central depigmentation, atrophy and well-defined hyperpigmented borders suggestive of DLE; b) Right buccal mucosa showing erosive OLP; c) Left buccal mucosa showing OLP with superficial candidiasis.

A dermoscopy was performed from the right cheek which was suggestive of DLE, showing the following features: prominent follicular openings with perifollicular scaling and erythema, diffuse superficial scaling, 'white rosettes', peppery brown pigmentation, pinkish white background of erythema and white structureless areas [Table/Fig-2]. Complete blood counts, erythrocyte sedimentation rate, liver function tests, renal function tests were done for the patient which were normal. Furthermore, to confirm the suspicion of DLE, a skin biopsy was performed two weeks after the first visit, which showed: hyperkeratosis, follicular plugging, moderate acanthosis, basal cell degeneration and patchy lympho-histiocytic infiltrate in the upper dermis [Table/Fig-3a,b]. These features were therefore conclusive of DLE. Antinuclear antibody test was done in view of DLE and it was negative.

The patient was seronegative for Human Immunodeficiency Virus (HIV), Hepatitis B surface antigen (HBsAg) and Hepatitis C Virus (HCV). The lesions over the left buccal mucosa were suspected to be leukoplakia and a biopsy from the patch was performed. It showed features of squamous cell carcinoma: dysplastic epithelium



[Table/Fig-2]: Dermoscopy of facial plaque: Black circles- white rosettes; yellow circles- prominent follicular openings; black stars- perifollicular scaling; blue stars- pinkish white background of erythema; blue arrows-white structureless areas; green arrows-peppery brown pigmentation.



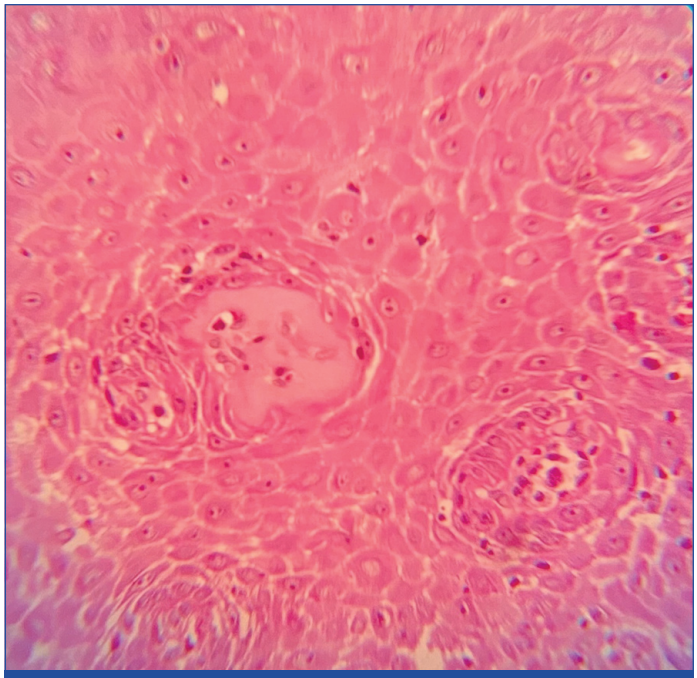
[Table/Fig-3]: Histopathology of facial DLE: a) (H&E; 100×) Follicular plugging (black arrow), irregular acanthosis, basal cell degeneration (blue arrow) and patchy inflammatory infiltrate; b) (H&E; 100×) Prominent basal cell degeneration with lymphocytic infiltrate (blue arrow).

with cellular and nuclear pleomorphism, nuclear hyperchromatism, individual cell keratinisation, loss of cohesion, altered mitotic activity and intra epithelial keratin pearl formation [Table/Fig-4]. However, for the violaceous lesions over the bilateral buccal mucosa, it was suspected OLP (erosive type) and a biopsy was performed from the right buccal mucosa which showed following features: hyperkeratosis, parakeratosis, spongiosis with irregular acanthosis, marked basal cell degeneration with a band like lymphocytic infiltrate in the upper dermis [Table/Fig-5]. These histopathological findings favoured OLP. A Direct Immunofluorescence (DIF) from the right buccal mucosa for IgG, Immunoglobulin G (IgG) and C3 was negative, which confirmed the oral lesion to be LP and not DLE with oral involvement. With this, it was concluded as a diagnosis of SCC invoving left buccal mucosa, bilateral OLP and DLE on the external skin, i.e., right cheek, lips.

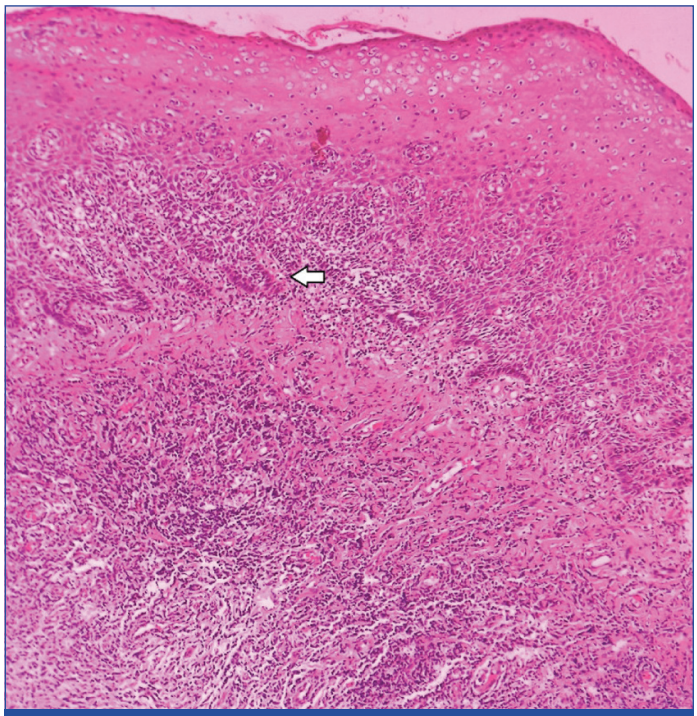
For the facial plaque, the patient was started on topical clobetasol propionate 0.05% ointment and systemic hydroxychloroquine 200 mg once daily. The OLP lesions were treated with alternate day applications of triamcinolone acetonide and tacrolimus. For the OSCC, patient was referred to the surgical oncologist who performed a wide local excision with split thickness skin graft and sentinel node biopsy followed by supra omohyoid neck dissection. Postoperatively, patient was given adjuvant radiotherapy. Four months after radiotherapy, the patient was comfortable and had a scar over the left buccal mucosa. The patient is still under treatment for DLE and OLP which shows mild improvement.

DISCUSSION

Discoid lupus erythematosus is the most common subset of chronic cutaneous LE. Photosensitivity, trauma, drugs, infections etc., can exacerbate lesions. Atrophy, scarring and central depigmentation



[Table/Fig-4]: Histopathology of SCC (H&E; 100x) Cellular and nuclear pleomorphism, altered mitotic activity and intraepithelial keratin pearl formation.



[Table/Fig-5]: Histopathology of OLP (H&E; 100×) Hyperkeratosis, parakeratosis, irregular acanthosiswith basal layer degeneration and a band-like inflammatory in the upper dermis infiltrate (white arrow) with exocytosis.

are key diagnostic features [1]. OLP presents as whitish streaks forming a lace like network on the buccal mucosa. Several clinical types have been reported viz., reticular, bullous, erosive, atrophic and plaque like [2].

Since the first case reported in 1977, LE-LP overlap syndrome has been a topic of deliberation [3]. The histological and immunological features of this entity blend with both diseases involved, thereby creating a quandary. The terms 'co-existence' and 'overlap' may create confusion and need to be delineated clearly [4]. Some reports have suggested a high incidence of Oral Squamous Cell Carcinoma (OSCC) in OLP patients and have implicated OLP as a premalignant lesion [4]. Patients with OLP, particularly the erosive type, have an increased incidence of OSCC development and should be monitored closely [5]. Lupus Erythematosus-Lichen Planus overlap syndrome (also known as Lupus Planus) was first proposed in 1977 by Romero and later established by Nagao K

and Chen KR [6,7]. Current criterion for 'true overlap' necessitates histological and immunological features of both LE and LP to be found in conjunction in the same tissue specimen. If not so, the term 'co-existence' is more apt [6].

Lupus Erythematosus-Lichen Planus overlap has been reported in approximately 50 cases in the past [7]. The DIF shows presence of IgG, granular C3 and IgM deposits (suggestive of LE) and cytoid body staining with shaggy fibrinogen along the basement membrane zone (suggestive of LP) [7]. Early DLE dermoscopically presents as erythema, telangiectasias, diffuse white scaling, keratotic follicular plugging, perifollicular halo etc., late lesions present as white structureless areas, blue grey globules, dotted vessels etc. The presence of 'white rosettes' under polarised light due to hyperkeratosis of follicular infundibulum is considered as a specific clue to the diagnosis of DLE among all inflammatory dermatoses is also noted in the case being reported [8]. OLP can have a benign self-limiting course to more florid manifestations deeply affecting patients' quality of life. The erosive form of OLP may give rise to malignancies [9].

Kiyani A et al., have reported a case of SLE with OLP (reticular type). The author has mentioned about the 'co-existence' of both conditions with negative DIF assay of the oral mucosa for IgG, IgM, IgA or C3. This goes against the criteria of true LE-LP overlap and thereby they have considered it as a synchronous presentation/co-existence [10]. This is similar to the present case where the DIF assay was negative with the additional finding of squamous cell carcinoma over the buccal mucosa.

Lupus Erythematosus and Lichen Planus are two distinct clinical entities. The autoimmune aetiopathology triggers the onset in both and thereby forms the link to 'overlap' or 'co-existence' as seen.

The main aetiological factors of OSCC are tobacco, alcohol, and emotional stress. Out of these factors, the patient described here presented chronic tobacco and alcohol consumption, which possibly influenced the occurrence of LP and the later occurrence of squamous cell carcinoma. Early detection of premalignant lesions and oral cancer is essential for achieving favourable prognosis [11]. Tobacco and smoking were found to be confounders in this case.

CONCLUSION(S)

In conclusion, considering that OLP is a relatively a common disease with malignant potential, the case highlights the need for early treatment and follow-up of precancerous oral lesions. This case of co-existing LE and LP in the same individual is being presented in order to highlight the importance of DIF technique to confirm such an association. Early oral biopsy is advised in all patients presenting with precancerous lesions such as leukoplakia, ulceration, erythroplakia, or LP in order to improve the quality of life.

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